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Bronchovenous Fistula During Adult Cardiac Surgery: A Case Report

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Bronchovenous fistula (BVF) associated with adult cardiac surgery is a rarely reported life-threatening condition. We present a 75-year-old woman who developed a BVF during cardiac surgery. Dense adhesion in the pleural and pericardial cavities was noted. Restrictive pulmonary pathology required high airway pressure. Transesophageal echocardiography and hemoglobin measurement were helpful for the timely diagnosis of BVF, which was controlled by transection of the right upper pulmonary vein where a vent catheter had been inserted. Injuries around the cannulated site presumably initiated the BVF, which was worsened by high-pressure ventilation. Therefore, cannulation site might be a risk factor for BVF. (A&A Practice. 2020;14:e01321.)

GLOSSARY

BVF = bronchovenous fistula; **ECMO** = extracorporeal membrane oxygenation; **EQUATOR** = Enhancing the Quality and Transparency Of health Research; **I:E** = inspiration/expiration; **IVC** = inferior vena cava; **LA** = left atrium; **LV** = left ventricle; **PEEP** = positive end-expiratory pressure; **PIP** = peak inspiratory pressure; **RUPV** = right upper pulmonary vein; **SAE** = systemic air embolism; **TEE** = transesophageal echocardiogram

Bronchovenous fistula (BVF) is mainly reported in neonates with respiratory diseases, including respiratory distress syndrome, or in adults with traumatic lung injuries. BVF causes systemic air embolism (SAE), resulting in myocardial or cerebral infarction, with a mortality rate of 48%–80%.[1] However, since there have been few reports of BVF associated with cardiac surgery in adults,[2,3] the pathophysiology and strategy for BVF during cardiac surgery in adults are unclear.

We present an adult patient who developed BVF with SAE during cardiac surgery. We discuss the possible etiology of BVF, contributing risk factors, and diagnostic measures adopted in this case. We obtained written consent for publication from the patient's spouse. This manuscript adheres to the applicable Enhancing the Quality and Transparency Of health Research (EQUATOR) guidelines.

DESCRIPTION

A 75-year-old woman with chronic mitral regurgitation, tricuspid regurgitation, and atrial fibrillation presented for elective mitral valve replacement, tricuspid valve annuloplasty, and maze procedure. Her medical history included breast cancer, treated with left mastectomy and radiation, and rheumatoid arthritis. She received home oxygen therapy of 0.5 L/min for chronic heart failure.

Preoperative transthoracic echocardiogram revealed preserved left ventricular (LV) ejection fraction (65%) with severe mitral and mild tricuspid regurgitation. Spirometry showed restrictive lung dysfunction (% vital capacity, 22.8%; forced expiratory volume in 1 second, 86.4%). A chest x-ray showed moderate bilateral pleural effusion. Her hemoglobin level was 10.4 g/dL.

We induced general anesthesia and inserted central venous and pulmonary artery catheters. Due to restrictive lung dysfunction and pleural effusion, the patient required high peak inspiratory pressure (PIP) of 30 cm H_2O to maintain an adequate tidal volume (150–200 mL). The Inspiration:Expiration (I:E) ratio was 1:1.5. The positive end-expiratory pressure (PEEP) was initially 5 cm H_2O.

Intraoperatively, we observed dense tissue adhesion and high tissue fragility, especially in the pleural and pericardial cavities. The patient was placed on cardiopulmonary bypass following aortic and right atrial cannulations and her heart was arrested. We inserted a vent catheter into the left atrium (LA) via the right upper pulmonary vein (RUPV) after exposing the RUPV in the dense pleural adhesion. We replaced the mitral valve with On-X 25 mm (On-X Life Technologies, Austin, TX) through a transseptal approach and repaired the tricuspid valve with Physio Tricupid ring 26 mm (Edwards Lifesciences, Irvine, CA). Immediately after the first aortic declamping, we observed bloody drainage in the endotracheal tube. We increased the PEEP from 5 to 15 cm H_2O to control this drainage.

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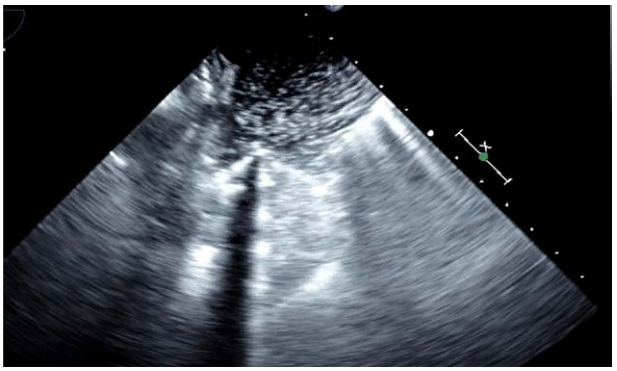

Figure. Transesophageal echocardiogram after the first aortic declamping. Midesophageal 2-chamber view demonstrating large amounts of air bubbles swirling in the left atrium.

At that time, 50 cm H_2O of PIP was required to maintain minute volume. Transesophageal echocardiogram (TEE) showed air bubbles swirling in the LA and LV. Electrocardiogram showed ST elevations in leads II and V_5. The patient soon developed ventricular fibrillation. Bleeding from the LA incision and the intrathoracic inferior vena cava (IVC) necessitated a second and third aortic cross-clamping, respectively. After the third aortic declamping, a large volume of bloody drainage spouted from the endotracheal tube, which was uncontrollable even with 15 cm H_2O PEEP. Blood gas analysis of the endotracheal drainage revealed a hemoglobin level of 9.6 g/dL, which was almost consistent with that of the blood collected from the arterial line. TEE showed continuous air bubbles coming into the LA that appeared in large volumes with each cycle of ventilation (Figure; Supplemental Digital Content, Video 1, http://links.lww.com/AACR/A376). Due to the uncontrolled endotracheal hemorrhage and systemic air entrainment, we promptly cross-clamped the aorta for the fourth time.

VIDEO+

The air entrainment visualized in the LA on TEE with mechanical inspiration and the endotracheal drainage hemoglobin level informed the diagnosis of BVF. Since the RUPV was cannulated for the LA vent, this seemed the most likely location, and the RUPV was excised. This led to a remarkable decrease in air entrainment and endotracheal hemorrhage. Low LV contractility (visual ejection fraction of 20%) without clear regional wall motion abnormalities and unstable hemodynamics prompted the initiation of an intra-aortic balloon pump and venoarterial extracorporeal membrane oxygenation (ECMO). We transferred her to the intensive care unit, without sternal closure, with mechanical circulatory support. The operative time was 630 minutes. The bloody drainage suctioned from the endotracheal tube was >2500 mL. On the seventh postoperative day, the patient underwent sternal closure. Due to her unstable hemodynamics, computer tomography imaging of her brain was deferred. However, comatose status, anisocoria, lack of spontaneous breathing, and absent extremity movement strongly suggested significant central nervous system damage. Because of multiple organ failure, we stopped ECMO with her family's consent and confirmed her death on the 12th postoperative day.

DISCUSSION

To date, only 2 BVF cases associated with cardiac surgery in adults have been reported.[2,3] The pathophysiology of BVF during adult cardiac surgery remains incompletely elucidated. Our patient developed a BVF during open cardiac surgery with concomitant SAE, including possible myocardial infarction and likely cerebral infarction, which caused an early death.

The BVF most likely developed during dissection of the adhesion around the RUPV for the vent catheter and widened during mechanical ventilation with high PIP. Previous reports showed that BVF is extrinsically traumatic or nontraumatic,[3] and both of these mechanisms were presumed in our case. The former mechanism is a procedure-related injury. During the repair, the surgeon pointed out that on the insertion of the vent catheter from the RUPV, he might have injured the right bronchial tree at the back of the vein with the scalpel. Tissue adhesion and fragility arising from a connective tissue disorder or postradiation therapy also contributed to damage in multiple locations, including the LA, IVC, and lung surface. The latter mechanism is overpressure injury, where alveoli rupture into small veins and capillaries.[2,4] This is mainly reported in neonates with respiratory diseases requiring high airway pressures. Similar overpressure injury possibly occurred in our case, because the airway had been exposed to 50 cm H_2O PIP after the first aortic declamping. This high PIP could have resulted in alveolar rupture, leading to BVF.

SAE is a leading cause of death after BVF. In our case, SAE involving major organs seemed to cause the patient's early death, as in previous reports.[2,3] The ST elevations in leads II and V_5 suggested air emboli in the right and left coronary arteries. Neurological findings indicated severe central nervous system injury, leading us to suspect cerebral air emboli and subsequent massive cerebral infarction. Given the high mortality associated with SAE, immediate suspicion of BVF and appropriate treatment are important. Hsaad et al[3] reported a case where a patient died intraoperatively and emphasized the usefulness of TEE for the diagnosis. In cardiac anesthesia, intracardiac air is very common after an aortic declamping. The important difference between usual residual intracardiac air and BVF is the continuous incoming LA air. If TEE shows continuous inflow of air bubbles into the LA, BVF should be suspected and immediate action should be taken for further SAE prevention.

In cardiac anesthesia, bloody or pinkish discharge in the endotracheal tube prompts anesthesiologists to consider the differential diagnoses: pulmonary edema, catheter-induced pulmonary artery perforation,[5] and BVF. The hemoglobin level of the discharge and the blood indicating that the discharge was mostly blood and the findings of TEE suggestive of bronchovenous communications enabled the timely recognition of BVF.

Once BVF is recognized, the next steps are prevention of further SAE development and definitive therapy for BVF. The suggested initial management for SAE includes placing the patient's head down, providing 100% oxygen, and decreasing the airway pressure.[6] Administration of 100% oxygen maximizes the patient's oxygenation and reduces the embolus volume by eliminating nitrogen.[7] A canine study showed that increased airway pressure enabled air to enter the pulmonary circulation.[1] Saada et al[8] reported a decrease of air bubbles into the LA on lowering airway pressure in chest trauma patients. Minimum tidal volume and airway pressure can be a therapeutic measure for BVF.

Selective ventilation of the noninvolved lung with a double-lumen tube or bronchial blocker is the optimal method of reducing airway pressure.[1,9] Reinhartz et al[6] successfully treated a neonate with BVF through 3-day selective ventilation. ECMO is another option to reduce airway pressure, especially if ventilation or hemodynamics is highly compromised.[3,6] The successful definitive therapy for BVF in our case was the extended dissection and transection of the cannulation site on the RUPV. After successful treatment of BVF, hyperbaric oxygen therapy could help to reduce the damage of SAE.[10,11] However, we did not proceed to the therapy because the patient was on mechanical circulatory support and too unstable to be transferred to the available facility. We had routinely inserted the vent tube via the RUPV for a clearer surgical field. Since dense adhesion dissection was the potential cause of the BVF, we now insert the vent catheter through a transseptal puncture from the right atrium when tissue adhesion is expected, such as in this case, or postthoracotomy.

BVF is a rare complication that most commonly affects neonates, but can also occur during cardiac surgery after traumatic LA vent insertion. TEE and continuous air entrainment in the LA help to diagnose BVF. Prompt recognition and treatment is imperative, because this condition is associated with high mortality due to SAE.

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